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Successful outcome in preeclamptic rudimentary horn pregnancy

Preeklamptik rudimenter boynuz gebeliğinde başarılı sonuç

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Abstract

Unicornuate uterus with rudimentary horn is an uncommon type of mullerian duct malformation associated with various gynecological and obstetrical complications. Rudimentary horn pregnancy is a rare entity and the majority have rupture of gravid horn leading to maternal and fetal morbidity and mortality. A case of rudimentary horn pregnancy at 32 weeks and 6days with pregnancy induced hypertension is reported where proper management results in successful pregnancy outcome. (J Turkish-German Gynecol Assoc 2011; 12: 53-5)

Key words: Rudimentary horn, unicornuate uterus, pregnancy, pre-

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Özet

Rudimenter boynuzlu unikornuat uterus, çeşitli jinekolojik ve obstetrik komplikasyonlarla ilişkili olan nadir bir müller kanal malformasyonudur. Rudimenter boynuz gebeliği nadir bir durumdur ve çoğunluğunda gebe boynuz rüptüre olarak maternal ve fetal morbidite ve mortaliteye neden olmaktadır. Uygun tedavinin gebeliğin başarılı şekilde sonuçlanmasını sağladığı 32 hafta ve 6 günlük rudimenter boynuz gebeliği ile gebeliğin neden olduğu hipertansiyonu olan bir olgu bildirilmektedir. (J Turkish-German Gynecol Assoc 2011; 12: 53-5)

Anahtar kelimeler: Rudimenter boynuz, Unikornuat uterus, Gebelik, Preeklampsi

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Introduction

Unicornuate uterus with rudimentary horn is an uncommon type of mullerian duct malformation. It is associated with various gynecological and obstetrical complications.

Rudimentary horn pregnancy is a rare entity, the incidence being 1 in 76,000 (1, 2). Very rarely, live births in rudimentary horn pregnancy is reported in the literature as the majority have rupture of the gravid horn, leading to extensive intraperitoneal hemorrhage and maternal and fetal morbidity and mortality. We present a case of rudimentary horn pregnancy with preeclampsia where proper management results in successful pregnancy outcome.

Case Report

A 21-year-old primigravida at 32 weeks and 6 days of gestation with preeclampsia was referred to the department of obstetrics and gynecology, BHU for decreased fetal movements for 1 day. She had had bilateral pedal edema for the previous 2 months. She was diagnosed as a case of preeclampsia and was on 500 mg of methyl-dopa three times daily for the previous 15 days. There was no history of dysmenorrhoea and her menstrual cycles were regular. She had received 2 doses of betamethasone. On examination, the patient's vital signs were stable. Her blood pressure was 160/100 mmHg.

Obstetrical examination revealed a fundal height corresponding to 32 weeks of gestation and deviation of the uterus to the right. The fetus was cephalic and fetal heart rate was 114 bpm. Cardiotocography was carried out and was nonreactive. Her hemoglobin was 12.5 gm/dl, urine albumin 2+, uric acid 7.2 mg/dl, creatinine 1.0 mg/dl and random blood sugar 69.0 mg/dl. Her liver function tests and renal function tests were normal. The ultrasonography (USG) of the second trimester done at the periphery was reported to be normal. Repeat USG with a biophysical profile (BPP) was done which revealed a 4/10 score with an amniotic fluid index of 8. The patient was moved for emergency caesarean section in view of low BPP (fetal distress). On laparotomy pregnancy was located in the right horn of uterus and the left horn was approximately 6 weeks size. The right horn was extensively fused to the left. The left and right fallopian tubes and ovaries were attached to the respective horns (Figure 1). Low transverse incision was made over the right horn and the baby was delivered. Placenta was delivered by control cord traction. Placenta and membranes were normal on gross examination. Clamps were applied and the right horn excised along with right salpingectomy (Figure 2). Both ovaries were preserved. The baby was a 1560 gm female with an Apgar score of 8/10 and 9/10 at 1 min and 5 min respectively.

The patient made an uneventful recovery and was discharged home on the tenth day. The follow up at 6 weeks was normal.

Discussion

Mullerian duct anomalies are infrequently encountered in day to day gynecological practice. The incidence of mullerian duct malformation in the general population is estimated to be 4.3% (3). The unicornuate uterus with a rudimentary horn is a rare type of mullerian duct malformation and results from the defective fusion of the malformed duct with the contra-lateral duct, the incidence being 0.4% (3). In 80-90% of the cases rudimentary horn is not connected to the uterine cavity (4).

Rudimentary horn pregnancy (RHP) is a very rare entity. Conception occurs in the rudimentary horn due to transperitoneal migration of sperm or zygote, although a corpus luteum has been observed on the contralateral side in only 10% of the cases (5). Rudimentary horn pregnancy has a very poor maternal and fetal outcome. The most common and life threatening complication of rudimentary horn pregnancy is rupture (80%). The rupture usually occurs in the first and second trimester (80%) and infrequently in the third trimester (20%) (6). Rupture leads to torrential intraperitoneal bleeding with severe maternal and fetal mortality. Here lies the importance of prerupture diagnosis but the prerupture diagnosis is extremely difficult. Literature shows a preclinical detection rate of 8% and a preoperative detection rate of 29% only (7). The diagnosis is possible only with a high index of clinical suspicion and radiological investigations (USG & MRI) (7, 8). The bimanual finding of palpable mass extending outward from the uterine angle (Baart de la faille's sign) or displacement of the fundus to the contralateral side with rotation of the uterus and elevation of the affected horn (Ruge Simon Syndrome) or deviation of the uterus to one side with an adnexal mass in pregnancy should arouse suspicion of a rudimentary horn. USG in early trimesters are helpful in diagnosing a case of RHP. Tsafrir et al. suggested the following criteria for early sonographic diagnosis of rudimentary horn pregnancy (8). (i) a pseudo pattern of an asymmetrical bicornuate uterus, (ii) absent visual continuity in tissue surrounding the gestational sac and the uterine cervix and (iii) the presence of myometrial tissue surrounding the gestational sac. Additionally, MRI can be used to confirm the diagnosis before laparatomy is undertaken. Buntugu et al have also shown that in low resource centres, placement of a Foley catheter into the uterine cavity and performing a transabdominal ultrasound scan can conclusively exclude an intact uterine pregnancy (9).

In addition to rupture, the RHP usually lead to missed abortion or intrauterine death; and rarely, fetal survival has been reported. Until 1999, only 13 neonatal survivals were reported in the English Literature (10). In an exhaustive review of 20th century literature, Nahum reported only 6% of RHP which have progressed to term, with a 13% neonatal survival (6). Goel et al. have reported a case of post dated RHP but the baby could not survive following delivery (11).

Ours is a case of RHP referred to our hospital in the third trimester with successful fetal and maternal survival. The preoperative diagnosis was not possible in our case, as the early pregnancy USG was done at a peripheral hospital where there was no expertise and it is difficult to diagnose RHP in cases of late trimester USG.



Figure 1. Unicornuate uterus and rudimentary horn



Figure 2. Excised rudimentary horn

The endometrium of the rudimentary horn has been described as thinner than usual and sometimes even non functional (12). Pathological placentation may lead to placenta accrete, as reported in various cases (13, 14). It is also possible that poor placentation in horn pregnancy may be a cause of preeclampsia and fetal distress or low BPP in our case.

The management of RHP is laparotomy and surgical excision of the rudimentary horn and tube, as was done in our case. In the literature, the laparoscopic removal of pre and post rupture RHP is reported but this is carried out in cases of early diagnosis (6, 15).

Conclusion

RHP is an extremely rare entity. The high index of suspicion is important to diagnose it early i.e. before rupture, to prevent life threatening consequences. A timely and proper management can result in a successful pregnancy outcome even in horn pregnancy.

Abbreviations

PIH: Pregnancy induced hypertension, BPP: Biophysical profile, USG: Ultrasonography, RHP: Rudimentary horn pregnancy

Consent

Written informed consent was taken from the patient for the publication of this case report and any accompanying images. A copy of the written consent is available for the review of the Editor in Chief of this journal.

Conflict of interest

No conflict of interest is declared by authors.

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